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# Two cases of concha bullosa in a contemporary Cypriot skeletal collection

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**Abstract**: *Concha bullosa is the hypertrophy of the superior, middle, or inferior nasal conchae, most commonly referring to the pneumatisation of the middle conchae. It is considered to be the most common anatomical variant of the osteomeatal complex, rather than a pathological development. Though it is common, its aetiology is poorly understood. It is unclear whether sex or ethnicity impacts on the prevalence of concha bullosa, though some research suggests a correlation. Some researchers have argued that concha bullosa predisposes individuals to sinusitis, but the link is not consistent.*

*In this paper, the authors present two new skeletal cases of bilateral concha bullosa identified in female individuals taken from the Cyprus Reference Research Collection (CRRC). This work aims to highlight the limitations associated with the palaeopathological diagnosis of inflammation and the interpretation of skeletal lesions that may be related to sinusitis or infection of the osteomeatal complex in archaeological bone, in relation to the presence of concha bullosa.*

**Key words**: inflammation; infectious disease; osteomeatal complex; palaeopathology; bioarchaeology; sinusitis

## Introduction

The nasal conchae, or "turbinates," are three shell-like structures contained within the nasal cavity. The two bones are arranged vertically and defined as superior, middle/intermediate, and inferior; the inferior nasal conchae are the largest of the three (Gray & Goss 1974). The superior and intermediate conchae are bilateral inferior

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projections of the lateral mass of the ethmoid bone. The inferior turbinates are a separate structure located on the lateral nasal wall and they are articulated medially with the vomer. The intermediate conchae do not normally contain air cells; however, in certain instances they can become pneumatised, causing hypertrophy of the conchae leading to concha bullosa.

Concha bullosa is defined by Stallman et al. (2004:1615) as "a middle turbinate with pneumatization extending caudally at least 50% of the vertical height of the middle turbinate". Concha bullosa was first noted by Zuckerkandl (1893) and is considered one of the most common sinonasal anatomical variations (Yiğit et al. 2010; Zuckerkandl 1893). Development of concha bullosa is argued to be linked to trauma to the nasal cartridge during growth and/or to genetic factors (Al-Qudah 2008; Haruna et al. 2005). A study conducted with twins by Chaiyasate et al. (2007) found no significant variation between the paranasal sinuses in the two siblings, suggesting that the development of concha bullosa may also be affected by environmental factors.

Within the literature there is a lack of standardisation in the diagnosis of concha bullosa (Ahmed et al. 2018), which may lead to the over- or under-representation of this anatomical variant within a population (Stallman et al. 2004). Some papers base their diagnosis on any aeration of the conchae even if it does not impact the bulbous portion (Smith et al. 2010; Tiwari & Goyal 2018) whilst others only provide a diagnosis if the patient demonstrates aeration moving caudally into the bulbous portion of the middle concha (Zinreich et al. 1988). Concha bullosa is commonly classified as one of three types (Bhatt 1997; Bolger et al. 1991; Hatipoğlu et al. 2005):

1. lamellar – vertical lamella is pneumatised;

2. bulbous – inferior portion of the concha is pneumatised;

3. extensive – the vertical lamella and inferior concha are pneumatised.

In clinical settings, concha bullosa is often asymptomatic in its early stages, and is commonly discovered during other medical procedures. It can become clinically significant if the conchal enlargement narrows the nasal meatus obstructing nasal flow (Erkan et al. 2017). Concha bullosa may require surgical intervention to remove or reduce the affected concha or conchae. If necessary, the septum may require surgical intervention to correct any deviation that may have resulted from the pneumatization of the concha (Perić et al. 2009).

Concha bullosa is not considered pathological but it is rather considered an anatomical variant with some clinical significance with regard to the epidemiology of inflammation and infection that may affect the osteomeatal complex (Earwaker 1993). The osteomeatal complex acts as an anatomical channel which links the frontal sinuses, anterior ethmoid air cells and maxillary sinuses to the middle meatus, thus allowing for airflow and mucociliary drainage to occur (Tsai et al. 2006). Therefore, obstruction of ventilation and drainage of these units can consequently result in sinusitis (Khojastepour et al. 2015), or more generally speaking, upper respiratory tract diseases (Alkire & Bhattacharyya 2010). An enlarged concha is likely to inhibit ample airflow from entering and/or causing the septum to deviate. As such, the clinical significance of this skeletal feature relates directly to its obstructive nature, which has been considered a potential contributing factor in the presence of chronic sinusitis (Lam et al. 1996; Maru & Gupta 1999; Stallman et al. 2004; Unlü et al. 1994). In a sample of 54 patients, Aramani and colleagues (2014) found that 53.7% of patients with chronic sinusitis had two or more anatomical variations associated with the ostiomeatal system, whereas 33.3% of the cases had a single anatomical variation (Aramani et al. 2014). Individuals with chronic rhinosinusitis had at least two anatomical variants. They also found that deviated septum was the most common, followed by bilateral or unilateral presence of concha bullosa (Aramani et al. 2014).

It is debated whether the presence of concha bullosa can predispose individuals to infection (Calhoun et al. 1991; Clark et al. 1989). Most cases of concha bullosa in living individuals are found incidentally when a patient receives medical treatment for a sinus infection, suggesting correlation rather than causation (Lloyd 1990). Various studies have found concha bullosa in 48.5–64% of patients presenting with other sinus-related conditions or headache (Bolger et al. 1991; Hatipoğlu et al. 2005; Huang et al. 2008). There are, however, other studies that found no correlation between concha bullosa and sinus infections (Alkire & Bhattacharyya 2010; Cho et al. 2011; Stallman et al. 2004; Tonai & Baba 1996).

Although largely mentioned in the clinical literature, only a few studies refer to the condition in archaeological populations (**Table 1**). This is unlikely to be because the condition did not affect ancient populations, but most likely because the condition is rarely observed due to the fragility of the structures of the nasal cavity (Tonina et al. 2018). The fragile nature of these skeletal structures predisposes them to higher risks of taphonomic and excavation damage, and as such, they are rarely preserved intact, or if at all, to be examined (Magalhães et al. 2017; Mays et al. 2011). The oldest cases of concha bullosa come from the Iranian site of Tepe Hissar, ca. 3500–3000 BCE, and the Egyptian necropolis of Sheikh-Abd-el-Gurna, 11$^{th}$–20$^{th}$ Dynasty (Hagedorn et al. 2002).

The research presented herein documents the presence of two cases of concha bullosa discovered in a set of 20$^{th}$/21$^{st}$ century Greek Cypriot remains, presents a differential diagnosis of the condition, and discusses the limitations for palaeopathological diagnoses of sinusitis.

**Table 1.** Recorded cases of concha bullosa in the palaeopathological and archaeological literature. N/A denotes that the information could not be obtained.

| Location | Chronology | Sample size | No. of cases | Age | Sex | Reference |
|---|---|---|---|---|---|---|
| Tepe Hissar (Damghan, Iran) | 3500–3000 BCE | 184 | 1 | adult | female | Krogman 1940 |
| Kivutkalns (Island of Doles, Latvia) | 1800–500 BCE (Bronze Age) | 230 | 10 | adults | 1 female, 1 unkn. | Derums 1987 |
| Vučedolm (East Slavonia, Croatia) | 3000 BCE | 8 | 3 | adults | females | Hincak et al. 2013 |
| Castellón Alto (Granada, Spain) | 1900–1600 BCE | 53 | 1 | 50+ | female | Salvador et al. 2019 |
| Sheikh-Abd-el-Gurna (Thebes, Egypt) | 2024–1070 BCE (11th–20th Dynasty) | 273 | 10% | adults | males/ females | Hagedorn et al. 2002 |
| Barcelona, Spain | 500–1500 CE (Medieval) | N/A | 1 | adult | female | Campillo 2005 |
| Huntingdon, England | 1066–1485 (Medieval) | 55 | 1 | 50 | female | Mays et al. 2011 |
| Church of Saints Peter and Paul of Brentonico (Italy) | 8th–13th c. | 22 | 1 | c. 35 | male | Tonina et al. 2018 |
| Hospital of St. John the Evangelist (Cambridge, London) | mid-13th c. | 395 | 1 | adult | male | Cessford 2015 |
| Wharram Percy, England | 12th–14th c. | 687 | 17 | adults | 10 males, 7 females | Mays et al. 2014 |
| St. Nicholas's Church (Głogów, Poland) | 13th–14th c. | 102 | 1 | 40 | male | Kwiatkowska et al. 2011 |
| South Dakota, USA | c. 1650–1700 | N/A | 1 | 35 | female | Gregg 1988 |
| Évora Inquisition Court (Portugal) | 16th–17th c. | 7 | 1 | ? | unknown | Magalhaes & Santos 2013 |
| Copenhagen, Denmark | 16th–20th c. | 854 | 20 | 18 ad 2 sub. | 15 fem., 3 males | Anthony et al. 2011 |
| St. Pancras (London, England) | 19th c. | N/A | 1 | c. 35 | male | Emery & Woodridge 2011 |
| Tomersdorf-Toporow (Zgorzelec County, Germany) | 19th–20th c. | 32 | 2 | adults | females | Gawlikowska-Sroka et al. 2016 |

## Materials and methods

A total of 34 skeletons from the Cyprus Research Reference Collection (CRRC) were randomly selected for cleaning and inventorying in the collection's digital database as part of an ongoing project to archive the CRRC. During this process two female skeletons presenting with concha bullosa were identified. The CRRC is a contemporary skeletal collection that consists of 2000 skeletons dating from between 1973–2015. The collection is stored in the ossuary of St. Nicholas cemetery, located in Limassol, Cyprus, and is managed by the local Greek-Orthodox Church authorities. The CRRC consists of individuals once buried within the Greek-Cypriot cemetery of

St. Nicholas, who were later exhumed and placed in a communal ossuary. Exhumation is permitted by the Church in order to allow for for additional interments within the grave. Graves within this context are typically family-owned and may receive up to six interments over time. Once this number is exceeded, families can either purchase another grave plot or exhume the initially interred individuals and place them into the ossuary. Individuals are buried in extended, supine position with the arms on their side or crossed over the pelvis. Secondary interments are placed at the feet of the individual, and they are often kept in white pillowcases, more recently in white cloth sacks, or more rarely so, in plastic bags, wooden or metallic boxes.

The sex, age-at-death and ethnicity of the skeletons were obtained from cemetery records. Sex and age-at-death were confirmed by means of osteological analysis by examining skull and pelvic areas of the skeletons using standard morphological methods (Brooks & Suchey 1990; Buckberry & Chamberlain 2002; Phenice 1969; Walker 2008).

The presence of inflammation and possible infection of the structures of the osteomeatal complex, more specifically maxillary sinusitis, was assessed following generally accepted macroscopic criteria comprising pitting, erosion and new periosteal bone formation at and around the sinuses, as well as the maxilla and ribs (Roberts 2007). The nasal cavity was examined for spicules that may suggest a form of chronic infection, which could be secondary to maxillary sinusitis (Lee et al. 2008; Wells 1977). Dental pathology may also cause secondary sinusitis, however both individuals were edentulous, therefore the presence of dental pathology could not be observed.

Access to radiography was not possible currently due to access restrictions to the remains.

## Case presentation

The first case presented here is that of a Greek-Cypriot female, age 86 (ED16 SK79). The remains were in an exceptionally good state of preservation and almost complete, missing only some of the hand bones. The skeleton presented with bilateral concha bullosa (**Figure 1**). The left concha was significantly larger than the right, with the nasal septum presenting with a contralateral deviation to the right. The concha bullosa was graded using the criteria suggested by Bolger and colleagues (1991), with both conchae scoring 2 (bulbous), meaning only the inferior section of the conchae are pneumatised. Additional skeletal pathological findings include occipito-cervical synostosis or fusion of the first cervical vertebrae to the occipital bone (Mudaliar et al. 2013); a button osteoma on the right parietal bone; perimortem trauma to the lower limb, and a femoral plate inserted into the right femur to stabilise a fracture to the femoral neck.

The second case presented here is that of a Greek-Cypriot female, age 73 (SK RPT3). The remains were well preserved and relatively complete, though there was slight damage to the anterior portion of the vomer, and all carpals were missing along with most of the hand phalanges. This case also presented with bilateral concha bullosa with little to no deviation of the nasal septum (**Figure 2**). The left and right conchae were both graded 2 (bulbous type) based on Bolger and colleagues (1991), as only the inferior portion of the concha was pneumatised. The left concha was dominant, though macroscopically there was little difference between them. There were no other major pathological findings observed, except for normal joint degeneration relating to common osteoarthritis.

In both individuals there were no other signs of the superior or inferior nasal conchae being pneumatised. There were no signs of spicules or other bony growths within the nasal cavity. However, evaluation of internal skeletal structures using endoscopy or digital imaging such as radiography and computer tomography (CT) scans for example could provide a better method for assessing the presence and morphology of lesions endocranially.

All attempts to classify a skeletal anomaly as pathological must undergo a rigorous process of differential diagnosis (Pinhasi & Mays 2008). The differential diagnosis for concha bullosa as it relates to the presence of the condition in this suite of remains

**Figure 1**. Bulbous type bilateral concha bullosa and a contralateral deviated septum away from the dominant conchae in Case 1 – ED16 SK79.

**Table 2.** Differential diagnoses for concha bullosa and associated literature.

| Pathology | Similarities | Differences | References |
|---|---|---|---|
| Craniofacial fibrous dysplasia | Expansive swelling of middle nasal conchae. | Bone has a fibrous, "ground glass" appearance. Sclerotic margins. Very rare, only two noted cases in English literature. | Lisle et al. 2008; Saetti et al. 2004 |
| Ossifying fibroma | Asymptomatic, expansive swelling of turbinate | Non-pneumatised, early-stage: bony rim surrounding soft tissue | Salina et al. 2017 |
| Concha bullosa | Swelling/hypertrophy of middle nasal concha. Pneumatisation of the turbinate | N/A | Araújo & Magalhães 2013; Mays et al. 2011 |

is shown in **Table 2**. This process ruled out any other pathological conditions that present with a similar skeletal morphology.

Both cases were adult females of a similar age and ethnicity. ED16 SK79 appeared to present with a more advanced stage of concha bullosa compared to SK RPT3, based

**Figure 2.** Bulbous type bilateral concha bullosa in Case 2 – RPT 3.

on the increased volume of the nasal conchae, although both cases were graded as a 2 following Bolger and colleagues (1991). However, the rate of growth or pneumatisation of conchae bullosa is unknown, and therefore it is not possible to determine in which one of the two individuals the condition developed first, especially in the absence of direct medical records reporting the presence and progress of the condition. Furthermore, in the absence of relevant medical records, it is not possible to ascertain how long the individuals lived with this anatomical variant and what may have been the clinical consequences, if any.

Despite the small sample size, given that both skeletons with concha bullosa were females, one question that arises is whether sex plays a role in the development of the condition. There is conflicting evidence of whether intrinsic sex factors can influence the risk of developing concha bullosa (Prasad et al. 2016; Smith et al. 2010; Subramanian et al. 2005). Subramanian and colleagues (2005) found that females present with a higher prevalence of concha bullosa, whilst Smith and colleagues (2010) and Prasad and colleagues (2016) found no significant relation between sex and the development of concha bullosa. Similarly, Mays and colleagues (2014) found no statistical differences in the male-female prevalence of the condition. Clinical studies present with varied male-female differences ranging between higher prevalence in females (Koo et al. 2017; Smith et al. 2010), higher bilateral prevalence in males (Al-Rawi et al. 2019), and uniformity of the trait in both sexes (El-Din et al. 2021). This variability confirms the nature of concha bullosa as an anatomical variant, that may also be population specific.

It has been proposed that the prevalence of concha bullosa is higher in populations from temperate and warm climates (Koertvelyessy 1972). However high prevalence was also observed in populations from cold climates (Earwaker 1993), thus suggesting that the natural environment may not be the underlying causal factor (Gawlikowska-Sroka et al. 2016). Environmental and biological factors combined determine the external morphology of the nasal cavity, which throughout human evolution, it shows a significant degree of variability between human groups living in cold or warm environments (Carey & Steegmann Jr 1981; Davies 1932). Environmental stimuli are likely to further affect the nasal and paranasal structures, but not necessarily result in the pneumatisation of the concha. Similarly, septal deviation can also be a common finding in given population, however it may or may not be present in cases where pneumatization affects only one of the two concha (Mohebbi et al. 2012). This indicates that in addition to climate, concha bullosa and septal deviation can be population specific anatomical variants without environmental predisposition but rather variants of genetic and biological origin.

Although it is accepted that concha bullosa is a pathological condition per se, its presence could have a clinical significance because it can contribute to the development of inflammation or infection of the osteomeatal structures, such as chronic sinusitis. Because the comorbidity of concha bullosa's with other pathological conditions is known in the clinical literature, the majority of bioarchaeological accounts seek to establish the same relationships. The aetiology of sinusitis is multifactorial, and the presence of concha bullosa (with or without septal deviation) is only a contributing factor to the development of pathology.

Sinusitis can be primary or secondary. Primary sinusitis can be attributed to the living conditions, occupational and cultural practices of a population. Lewis (2002) and Roberts (2007) suggested that the presence of sinusitis in a population is highly related to bad air quality attributed to the growing industrialization of occupational tasks ranging from pottery kilns to metalworking and mining, but also to cultural and subsistence activities such as sitting around open fires and smoking (Lewis 2002; Roberts 2007). Secondary sinusitis can occur from dental disease such as periapical abscesses and periodontal disease, the presence of polyps, mucous retention cysts, and various allergies, all of which could contribute to chronic infection of the upper respiratory track (Mohebbi et al. 2012; Patel & Ferguson 2012; Troeltzsch et al. 2015), Generally speaking, the blockage or restriction of air flow through the nasal cavity from anomalous anatomical structures such as pneumatized concha and/or septal deviation, as well as blockage of the osteomeatal complex from chronic infection, could contribute to the development of sinusitis (Mohebbi et al. 2012). Systemic stressors visible on bone or teeth such as periosteal new bone formation (indicative of inflammation or infection) (Roberts 2019) and linear enamel hypoplasia can also be indicative of the presence of general chronic systemic inflammation such as sinusitis (Liebe-Harkort 2012; Roberts 2007; Smith-Guzmán 2015).

In bioarchaeology, the diagnosis of sinusitis is a complex and difficult task which, in addition to the presence of skeletal lesions, also requires advanced technology such as endoscopy or digital imagining to observe endocranial structures. This is mainly because the primary diagnostic features of sinusitis are soft tissue related and, they are symptomatic to the living in ways that do not manifest on the skeleton (Slavin et al. 2005). It is not unlikely though that chronic occurrence of sinusitis could affect associated skeletal structures and in turn promote either osteoclastic or osteoblastic activity in the sinuses, maxilla, and ribs (Fleming 1954; Roberts 2007; Tovi et al. 1992). Osteitis of the paranasal sinuses is the result of chronic inflammation residing within these bony structures (Georgalas et al. 2010; Melén et al. 1986) Similarly, bone remodelling and erosion may result from chronic inflammation from nasal polyps, which although they are benign soft tissue growths, they have been associated with bone remodelling and resorption (London & Reh 2016).

Although there is a direct relationship between these changes and sinusitis in the clinical literature, when erosive and proliferative lesions are found outside the paranasal sinuses, these may have a different pathophysiology, which may be other than sinusitis. Research on skeletal remains strongly suggests that rib lesions may be indicative of nonspecific lower (pulmonary infection) or upper (sinusitis) respiratory diseases, mostly due to tuberculosis (Matos & Santos 2006). Although those lesions associated with well-defined infectious disease are least likely to be misinterpreted, the presence of new periosteal bone formation with pitting can be more difficult to specifically deduce. Scurvy is known to cause periosteal bone formation in the maxillary region for example (Geber & Murphy 2012). Sinusitis resulting from dental disease is almost an impossible connection to establish unless a fistula between the tooth structure and the sinus is seen (Roberts 2007). Yet, inflammation associated with antemortem tooth loss may result in the production of periosteal bone near and around the affected alveolus.

It is however possible that evidence of chronic sinus inflammation does appear on bone, and may be visible macroscopically, however evidence of these lesions may be lost in the burial environment, especially new periosteal bone being so fragile. Similarly, bone porosity in this case is often associated with more fragile skeletal elements such as the internal surfaces of the maxillary sinuses, the maxilla, and ribs (Roberts 2007), preservation of which is often poor in archaeological skeletal assemblages.

Similarly, radiography and Computed Tomography (CT) may also have both advantages and disadvantages in the diagnosis of the disease in skeletal remains. Mays and colleagues (2014) used radiographs to establish the presence of sinusitis and its relation to concha bullosa. They found a build-up of lamellar bone inside the sinuses, which in the absence of new woven bone, they attributed to chronic, healed, sinusitis (Mays et al. 2014). However, although lamellar bone is thick and radiopaque, new (fresh) deposits of woven bone are more radiolucent, therefore, it can be easily missed on radiographs. Studies show different results regarding the value of radiography as a diagnostic tool for chronic sinusitis, but also dispute the approach that lamellar bone is the only relevant change significant to the diagnosis of sinusitis (Laine et al. 1998; Stankiewicz & Chow 2002). Stankiewicz and Chow (2002) found 50% of patients with a strong history of chronic sinusitis had normal CT scans; therefore, the disease might be present but without any bone lesions. Using radiography, Laine and colleagues (1998) found that mucosal thickening was present in 98% of the patients examined, suggesting that other skeletal changes may occur. Therefore, unless erosion and bone proliferation are present and extensive (for the latter, multiple calcified deposits are present or lamellar bone), it is likely that these changes will not be visible on radiographs or CT scans. Thus, radiology may be of limited value to the researcher in determining, not only the presence of sinusitis, but also its healed or active character (Slavin et al. 2005).

## Conclusion

This study identified concha bullosa in two elderly female individuals. These individuals did not exhibit any other skeletal lesions that may be related to, or indicative of, sinusitis, inflammation or infection associated with the osteomeatal complex. This is, however, not conclusive of the presence or absence of osteomeatal-related pathology in these two individuals. This being a case of only two individuals, population references cannot be directly made at this point as more robust data sample is needed. Similarly, the Greek-Cypriot population remains largely understudied epidemiologically for such clinical conditions, which poses another limitation to our study.

Bioarchaeological references to concha bullosa remain rather limited. Although the identification of this trait may be easy on dry bone, interpretation of its presence and potential comorbidity or related pathogenesis to sinusitis remains challenging for bioarchaeologists. Despite being primarily considered an anatomical variant, consistent reporting of this trait, both in archaeological and clinical settings, may help shed more light on what the corresponding pathological implications of concha bullosa may be.

In the future, the authors aim to examine more skeletons from the Cyprus Research Reference Collection for the presence of concha bullosa and conduct a full radiographic study and, with the use of a digital endoscope, assess the internal structures of the maxillary sinuses. Additionally, the ongoing aim will be to also control for mastoiditis and dental disease, to better assess comorbidity and pathogenesis of concha bullosa.

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